Hadrotherapy Effectiveness in the Treatment of Mobile Spine Chordomas after Intralesional Excision and in the Management of Local Recurrences: A Retrospective Observational Study

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INTRODUCTION:

Chordoma is a malignant tumor with a mesenchymal nature, showing notochord differentiation. Some evidence suggests that it may originate from remnants of notochord cells located at the base of the skull, in the spine, and in the sacrococcygeal region.

It is a locally invasive tumor, and in a few cases it can spread to the lungs, to subcutaneous tissue, and lymph nodes. It is the most frequent primary malignant tumor of the spine (0.8 cases per 100,000). Ratio of male to female is 1,8/1. It most commonly affects the fifth and seventh decades of life.

METHODS: Retrospective observational study aimed at evaluating the outcomes in patients diagnosed with Chordoma or local recurrence of Chordoma, after surgical treatment with intralesional excision and implant of carbon fiber hardware, followed by hadrotherapy. Data were collected between January 2015 and April 2020. The study was carried on in three Institutions: the Spine Surgery Department of the Rizzoli Orthopedic Institute in Bologna, the National Center of Adrotherapy in Pavia and the Proton Therapy Center in Trento. Inclusion criteria were:

• patients aged 11 or over;

• patients diagnosed with Chordoma undergoing surgical treatment with inappropriate margins according to the Enneking classification;

• patients diagnosed with Chordoma of the mobile spine, whose localization and/or local extension was not suitable for en bloc resection surgery with marginal or wide margin according to the Enneking classification;

• patients diagnosed with local recurrence after en bloc resection.

RESULTS:

Twenty-one patients (14 M and 7 F) meeting the inclusion criteria were included in the study. The mean age was 61.4 years (range 12 - 81). The localization of the lesions was divided as follows: 3 (14%) in the cervical spine, 5 (24%) in the thoracic spine, and 13 (62%) in the lumbar spine.

Seven patients (33%) developed local recurrence after marginal resection. Local disease recurrence occurred at a mean of 52 months after surgery (range 20-94). These patients underwent intralesional excision of the recurrence and pedicle screw fixation with carbon hardware. Five patients already had anterior column reconstruction with a modular carbon prosthesis after en bloc resection. One patient had a homoplastic bone graft and one patient a homoplastic graft reinforced with a Harms cage.

Thirteen (62%) patients had undergone a previous intralesional surgery that was considered inappropriate, according to Enneking criteria. Among them, 2 underwent en bloc resection for T10 and T12 thoracic lesion respectively. Four patients underwent gross total excision, 5 underwent extracapsular intralesional excision of the lesion, while 2 had an intracapsular intralesional excision and excision. In patients undergoing en bloc resection, the anterior column was reconstructed with a modular carbon prosthesis for the T10 lesion and with a homoplastic graft for the T12 lesion. In 2 patients presenting with a cervical lesion, the anterior column was reconstructed with homoplastic bone graft and carbon plate. Since carbon fiber screws for lateral mass fixation were not available, posterior stabilization was achieved with using polyester sublaminar bands fixed with pre-shaped carbon rods. Among the 4 patients who underwent gross total excision, reconstruction of the anterior column was obtained with a homoplastic graft and carbon plate in 3 cases and with a modular carbon prosthesis in the last one. In 1 patient who underwent intracapsular intralesional excision, the anterior column was strengthened using PMMA. The remaining patients who underwent intralesional excision did not require anterior column reconstruction. All patients with a thoracolumbar localization, were stabilized posteriorly with carbon fiber pedicle screws.

Only one patient had never undergone any surgery for a C3 chordoma. He was then treated by extracapsular intralesional excision and reconstruction of the anterior column with homoplastic bone graft and carbon plate without any further posterior stabilization.

All patients underwent adjuvant hadrotherapy. Eight patients underwent carbon ion therapy with an average dosage of 68 GyRBE (range 54 – 73.6). One patient received a dose of 54 GyRBE due to previous photon irradiation at a dose of 54 GyRBE. Thirteen patients, on the other hand, presented an adjuvant treatment with Proton therapy. The mean dosage was 68.7 GyRBE (range 54-74). One patient received a dose of 56 GyRBE for a previous SBRT irradiation.

Four patients experienced a local recurrence with a mean interval of 21 months (range 16 - 27). Two patients had undergone en bloc resection with marginal margins at the beginning of their therapeutic workup and 2 patients had received an initial inappropriate intralesional procedure according to Enneking. Of these patients one died of disease (DOD) and 3 are alive with disease (AWD). The remaining 17 patients considered in the series have no evidence of disease (NED) at follow up. Of these patients, 2 presented musculoskeletal disease progression without any local recurrence.

The local disease control rate in the series considered was 71.8% at 24 months and 61.5% at 36 months.

DISCUSSION AND CONCLUSION: Hadrotherapy is able to obtain a local control rate of 71.8% at 24 months and of 61.5% at 36 months in the treatment of local Chordoma recurrences, after en block resection, after surgery with inappropriate resection margins when en bloc resection is not feasible because of the extension of the disease.